A FADED BEDTIME WITH RESPONSE COST PROTOCOL FOR TREATMENT OF MULTIPLE SLEEP PROBLEMS IN CHILDREN

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The sleep-wake cycles of 4 developmentally delayed individuals with longstanding severe sleep disturbances were regulated using a faded bedtime procedure with response cost. Bedtimes were systematically delayed for each individual, thus increasing the probability of short latency to sleep onset. The response cost component, consisting of removing the individual from bed for 1 hour, was implemented when an individual did not experience short latency to sleep onset. A fading procedure was then applied successfully to advance the bedtimes and to gradually increase durations of sleep. Specifically, all 4 individuals had decreased amounts of nighttime sleep that increased following treatment. Two of the 4 individuals showed excessive daytime sleep that decreased following treatment. Three of the 4 individuals experienced decreases in night wakings following treatment. Both environmental and biological manipulations of the sleep-wake cycle are hypothesized as mechanisms of treatment. The relative advantages of this procedure over other procedures for the treatment of pediatric sleep disorders are discussed, as are directions for future research.

DESCRIPTORS: pediatric sleep problems, developmentally delayed individuals, fading, response cost

Bedtime struggles or night wakings occur in approximately 20% to 30% of children between 1 and 4 years of age (Lozoff, Wolf, & Davis, 1985), and many children have both problems. Richman (1981) reported that 62% of children waking 5 or more nights per week had problems settling and long latency to sleep onset, and 35% of the waking group slept in the parents' room (a problem referred to as "cosleeping") on 3 or more nights per week. Estimates for cosleeping are as high as 70% of children with sleep problems (Lozoff et al., 1985). In addition to being very common and problematic for families, sleep problems tend to persist if untreated. Kataria, Swanson, and Trevathan (1987) found that 84% of children initially described by parents as having sleep problems continued to have problems at a 3-year follow-up.

Although treatments for insomnia have been experimentally validated in the adult literature (e.g., Borkovec, Grayson, O'Brien, & Weerts, 1979; Haynes, Price, & Simons, 1975; Haynes, Sides, & Lockwood, 1977), few empirical studies have documented the effectiveness of these interventions with

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children. Recommended treatments include door closing (Ferber, 1985; Wolf, Risley, & Meese, 1964), fading (Douglas & Richman, 1982; Ferber, 1985), reinforcement (Richman, Douglas, Hunt, Landsdown, & Levere, 1985), and relaxation and contingency contracting (Framer & Sanders, 1980). Much of the literature consists of anecdotal reports and case studies lacking in methodological rigor.

Notable exceptions are studies by Rickert and Johnson (1988) and Adams and Rickert (1989). Rickert and Johnson compared the efficacy of scheduled awakening with planned ignoring (extinction) for the treatment of night wakings. Scheduled awakening is a procedure in which the child is awakened prior to the expected occurrence of the spontaneous awakening. These scheduled awakenings are then gradually faded. Both treatments appeared to be equally effective in reducing night wakings; however, the planned ignoring treatment resulted in more side effects and was rated as less acceptable by parents. Adams and Rickert compared positive routines and graduated extinction in the treatment of bedtime tantrums. The positive routine treatment consisted of the implementation of several routines prior to the child's bedtime, setting an initial bedtime that was in accordance with the child's typical sleep onset time, and repeatedly returning the child to bed if he or she got out of bed. Both treatments were equally effective in reducing bedtime tantrums.

Scheduled awakening appears to be an effective, acceptable procedure for night wakings that does not produce substantial side effects. Graduated extinction and positive routines appear to be effective in treating bedtime tantrums. However, like the majority of interventions recommended for treatment of pediatric sleep problems, these procedures are designed to affect a single type of sleep problem. As previously noted, many sleep-disordered children show multiple sleep problems. Therefore, development and validation of a procedure that is relatively easy to use, has minimal side effects, and treats multiple sleep-related problems are warranted.

We have previously reported on a procedure involving bedtime fading that appeared to ameliorate multiple sleep problems for a 6-year-old girl with insomnia and attention deficit-hyperactivity disorder (AD-HD) (Piazza & Fisher, 1989). Similar to the procedure used by Milan, Mitchell, Berger, and Pierson (1981) and Adams and Rickert (1989), the treatment consisted of putting the child to bed at a time when sleep onset was highly probable. The bedtime was then gradually made earlier over time (i.e., fading). Several differences exist between the positive routines procedure and the bedtime fading procedure (Piazza & Fisher, 1989). First, in bedtime fading, the essential treatment component is hypothesized to be fading of the bedtime. In the positive routines procedure, several different responses on the part of the parent may affect the child's behavior (i.e., the routines, the fading, and/or the repeated returns to bed). Second, the bedtime fading procedure was used to affect a variety of sleep-related problems (e.g., delayed sleep onset, night or early wakings, short duration sleep). The dependent variable in the Adams and Rickert (1989) article was the bedtime tantrums, not the actual sleep of the child. Finally, the response to a child's tantrums in the fading procedure is to continue the baseline response (i.e., attend to him or her, allow him or her to get out of bed, have toys, etc.). The response of the parent to a child's continued tantrums in the positive routines procedure involves planned ignoring.

The purpose of the current investigation was to test further the efficacy of a faded bedtime procedure for the treatment of pediatric insomnia. To enhance treatment efficacy, a response cost component was added to the bedtime fading procedure. That is, it was hypothesized that adding a response cost would allow a more rapid fading of the bedtime. Finally, the current procedure takes into account developmental norms when assessing and treating a child's problematic sleep.

METHOD

Subjects

Subjects were 4 children referred for the assessment and treatment of self-injury to an inpatient unit that specializes in the treatment of mentally retarded children with severe behavior disorders. Although self-injury was the primary referring problem, the parents of all 4 individuals indicated that sleep problems were extremely disruptive to family life and that reducing these problems was a major goal of treatment. All 4 individuals were nonverbal, diagnosed as profoundly retarded, and met DSM-III-R criteria for insomnia. All parents indicated that supervision was necessary during night wakings; thus, not only were the individuals sleep deprived, but so were the parents. All individuals were free of medication for 2 weeks prior to and during the assessment and treatment of their sleep problems. Chronological ages of the individuals were Ann, 3 years; Mat, 4 years; Abe, 13 years; and Deb, 19 years.

Mat presented with delayed sleep onset, night wakings, and early wakings that had been present for at least 1 year. He also got in and out of bed frequently. This was a dangerous behavior because he would jump on top of the bedrails and off the bed without regard to the presence of hard objects or surfaces. During night wakings, he engaged in disruptive and destructive behavior (e.g., banging on walls, destroying furniture). Both Halcion and chloral hydrate had been tried without success to treat his sleep problems.

Ann exhibited delayed sleep onset, night wakings, early wakings, and excessive daytime sleep; these had occurred since 1 year of age. When she cried and engaged in self-injury during her night wakings, her parents brought her into bed with them. Halcion had been used unsuccessfully to improve Ann's sleep. For Ann, sleep treatment was initiated while she was an outpatient; follow-up data were collected 1 year later when she was admitted to the hospital for treatment of self-injury.

Deb was reported to have extremely variable durations of nighttime sleep (from 0 to 24 hr of sleep in a 24-hr period), a problem that had been occurring since age 10. She also had varying amounts of daytime sleep. Nights of short-duration sleep were characterized by delayed sleep onset and night and early wakings. Deb engaged in high rates of self-injury during her nighttime and early wakings. Halcion had been used to treat her sleep with limited success.

Abe also presented with delayed sleep onset and night and early wakings that had been present for at least 1 year. He typically engaged in disruptive and destructive behaviors during night wakings. In addition, he had successfully left the house on several occasions. Prior to admission, a trial of Halcion had no effect on Abe's sleep.

For Mat, Abe, and Deb, sleep treatment was initiated during their inpatient stay. Length of stay on the unit prior to treatment for sleep disorders was 23 days for Mat, 58 days for Abe, and 18 days for Deb.

Data Collection

On the inpatient unit, a momentary time-sampling procedure, in which the individual was scored as in bed or out of bed and awake or asleep at each half-hour interval by trained observers, was used continuously. If there was a question of whether the child was awake or asleep, the observer would stand within 1 ft of the child and whisper the child's name, using the absence of verbal or motor responses (e.g., opening eyes, vocalizing) as an indication that the child was asleep. In addition, a continuous frequency of in-and-out-of-bed behavior was scored for Mat for 3 days prior to and every

night following the introduction of treatment. The same 30-min momentary time-sampling procedure was used for Ann until the parents went to bed (at approximately 12 midnight). After their bedtime, the parents made two scheduled checks, one at 2:00 a.m. and one at 4:00 a.m. The parents were asked to record any other night wakings they observed.

Dependent Variables

Data are presented as a percentage of intervals of sleep that occurred during appropriate and inappropriate sleep times. Appropriate and inappropriate sleep intervals were established as follows. Developmental norms were used to determine the average number of hours of sleep required for each child (Weissbluth et al., 1981). Ideal sleep and wake times were then established using these norms in conjunction with the parental goals for their child's sleep and wake times. That is, each parent was informed of the average number of hours of sleep per day needed by an individual of their child's chronological age. The parents were then asked to state the time they would like their child to initiate sleep, awaken, and nap. This information was obtained after the children entered the hospital (for Mat, Deb, Abe) and just prior to treatment for Ann in order to prevent any changes in sleeprelated practices on the part of the parent as a result of the interview. The terms "appropriate" sleep and "inappropriate" sleep are used instead of "nighttime" sleep and "daytime" sleep to account for the fact that it is appropriate for children under the age of 4 to have some daytime sleep (i.e., naps). Thus, appropriate sleep time included day time sleep that occurred during regularly scheduled naps.

Percentage of intervals of appropriate sleep was defined as the number of sleep intervals occurring during the defined sleep period divided by the total number of intervals of the defined sleep period multiplied by 100. Percentage of intervals of inappropriate sleep was defined as the number of sleep intervals that occurred during the defined wake time divided by the total number of intervals of the defined wake time multiplied by 100. Frequency of night wakings was defined as the number of awake periods during the defined sleep times

that were both preceded and followed by a sleep episode of at least 15 min in duration.

Reliability

On the inpatient unit, interobserver agreement was assessed by having two independent observers score the child as awake or asleep. An occurrence agreement was scored if both observers scored the child as asleep. A nonoccurrence agreement was scored if both observers scored the child as awake. A disagreement was scored if one observer scored the child as awake and one observer scored the child as asleep. All reliability coefficients were calculated on an interval-by-interval basis. Occurrence agreement was calculated by dividing occurrence agreement by the sum of occurrence agreements and disagreements and multiplying by 100. Nonoccurrence agreement was calculated by dividing nonoccurrence agreements by the sum of nonoccurrence agreements and disagreements and multiplying by 100. Total reliability was calculated by dividing the sum of occurrence agreements and nonoccurrence agreements by the sum of occurrence agreements, nonoccurrence agreements, and disagreements and multiplying by 100. Reliability observations were conducted during both assessment and treatment on 23% of intervals for Mat, 33% of intervals for Deb, and 20% of intervals for Abe. Reliability observations were conducted on 31% of intervals for Ann during follow-up only. Mean interobserver occurrence agreement was 98% (range, 88 to 100) for Mat, 87% (range, 78 to 100) for Ann, 98% (range, 75 to 100) for Deb, and 93% (range, 67 to 100) for Abe. Mean nonoccurrence agreement was 90% (range, 0 to 100) for Mat, 96% (range, 92 to 100) for Ann, 99% (range, 88 to 100) for Deb, and 97% (range, 80 to 100) for Abe. Mean total reliability was 99% (range, 92 to 100) for Mat, 97% (range, 94 to 100) for Ann, 99% (range, 96 to 100) for Deb, and 98% (range, 92 to 100) for Abe.

Experimental Design

Treatment efficacy was assessed using a nonconcurrent multiple baseline across subjects design. The treatment package was introduced after 7 days of baseline for Mat, 11 days for Ann, 14 days for Deb, and 19 days for Abe.

Procedures

Baseline. During baseline, daytime sleep was interrupted for meals, medical procedures, therapy sessions, and necessary daily care. At other times sleep was neither encouraged nor discouraged. Night sleep was encouraged by prompting the child to go to bed at his or her bedtime following a consistent bedtime routine. This prompt was repeated once every 30 min if the child was out of bed. If early or night wakings occurred, the child was monitored by an adult and prompted to go back to bed at 30-min intervals. This baseline procedure was similar to the families' behavior at home; that is, all families indicated that they used a consistent bedtime routine with their child. In addition, all families indicated that, when their child awakened at night, they typically supervised him or her and periodically prompted the child to return to bed.

Treatment. The first step in treatment involved using baseline data to determine a bedtime at which rapid sleep onset (i.e., within 15 min) was highly probable. This initial bedtime (from which fading occurred) was determined by calculating the average sleep onset time during baseline and then adding 30 min (e.g., if the average time of sleep onset was 10:30 p.m. during baseline, then the initial bedtime was set at 11:00 p.m.). The child was not allowed to go to bed or fall asleep prior to this time or sleep past the scheduled wake time.

Fading consisted of adjusting the child's bedtime by 30 min each night based on latency to sleep onset for the previous night. If the child fell asleep within 15 min of bedtime, the bedtime was then made 30 min earlier on the next night. If the child did not initiate sleep within 15 min of bedtime, the bedtime was made 30 min later on the subsequent night.

The response cost component consisted of removing the child from bed and keeping him or her awake for 1 hr if sleep was not initiated within 15 min of bedtime. During this time, the child had access to the same items and attention as in baseline, but could not return to bed until 1 hr later. At the

end of the hour, the child was then put back to bed. This procedure was repeated throughout the night until the child was put to bed and sleep was initiated within 15 min.

To assess the effects of the procedure on other bedtime problems (e.g., night wakings and refusal to go to bed), the caretakers were instructed to continue responding to the child during night wakings or bedtime struggles as done during baseline. That is, if the child awakened during the night, the caretaker monitored the child and prompted him or her to go to bed at 30-min intervals. Ann's parents continued to bring her into bed with them.

The data on Deb's night wakings were possibly confounded when her physician initiated a nightly catheterization at 3:00 a.m., which coincided with her sleep treatment. Because she was not awakened and catheterized during baseline, night wakings were scored during the treatment phase if she did not reinitiate sleep within 15 min after the completion of catheterization.

Follow-up. Follow-up data were collected for Ann and Abe. Ann's data were collected on the inpatient unit 1 year after the treatment for sleep problems had begun at home. Abe's data were recorded by his mother 1 month postdischarge. Follow-up data were not available for Mat and Deb, and no information was obtained from their parents.

RESULTS

The percentage of intervals of appropriate sleep for each child is depicted in Figure 1. All children showed improvements in the percentage of intervals of appropriate sleep. Mat's sleep increased from an average of 78% in baseline to 87% after treatment; Ann averaged 75.8% of intervals of appropriate sleep in baseline, 89.2% after treatment, and 90% at 1-year follow-up. Deb averaged 57% of intervals of appropriate sleep in baseline and 72% after treatment. In addition, on 4 nights during baseline, her percentage of intervals of appropriate sleep was 0% and, in two of those 24-hr periods, she did not sleep at all. However, in treatment, she averaged

6 hr of sleep per 24-hr period, with all sleep occurring at appropriate times. Abe averaged 74% of intervals of appropriate sleep in baseline, 77% in treatment, and 86% at 1-month follow-up.

The percentage of intervals of inappropriate sleep is depicted in Figure 2. Ann's inappropriate (daytime) sleep decreased from an average of 11.3% of intervals in baseline to 2.1% of intervals after treatment and to 0.36% at 1-year follow-up. Deb averaged 9% of intervals of inappropriate sleep during baseline and 0% of intervals after treatment. Abe evidenced a small amount of inappropriate sleep (0.9%) in baseline, which was eliminated after treatment and at follow-up.

The frequency of night wakings is depicted in Figure 3. Three of the 4 individuals showed decreased night wakings following treatment. Mat averaged 0.3 wakings per night during baseline and 0 following treatment. For Ann, mean wakings per night decreased from 1.09 during baseline to 0.64 following treatment and decreased further to 0.09 at 1-year follow-up. Abe averaged 0.6 wakings per night during baseline, 0.2 during treatment and 0 at 1-month follow-up. The decrease in night wakings was marginal and probably not clinically significant for Deb (from an average of 0.3 wakings per night during baseline to 0.2 following treatment). The night wakings that occurred during treatment were almost exclusively when she was catheterized at 3:00 a.m. each night. Although she typically fell back to sleep fairly rapidly (within 15 min of the catheterization), on 3 nights following treatment she exceeded this 15-min criterion.

The frequency of Mat's climbing in and out of bed (Figure 4) decreased from a mean of 30 (range, 15 to 51) to low levels following treatment (M = 1.1; range, 0 to 20). Mat got out of bed on 8 of the 50 (16%) nights during treatment compared to 100% of the nights during baseline.

The percentage of nights when Ann was brought into her parents' bed (Figure 4) was quite high during baseline (M=84.3) but gradually decreased during treatment (M=45.4). At 1-year follow-up, her parents reported that they almost never brought the child into their bed (less than once every 2 months).

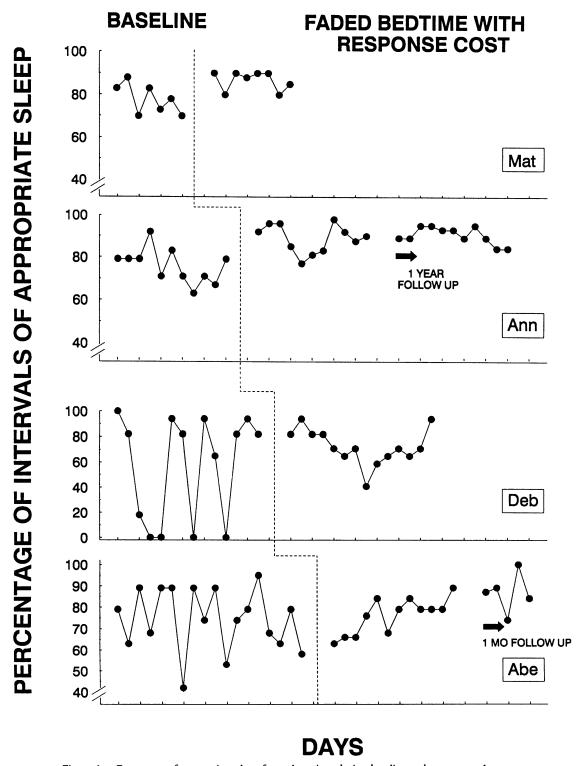


Figure 1. Percentage of appropriate sleep for each patient during baseline and treatment phases.

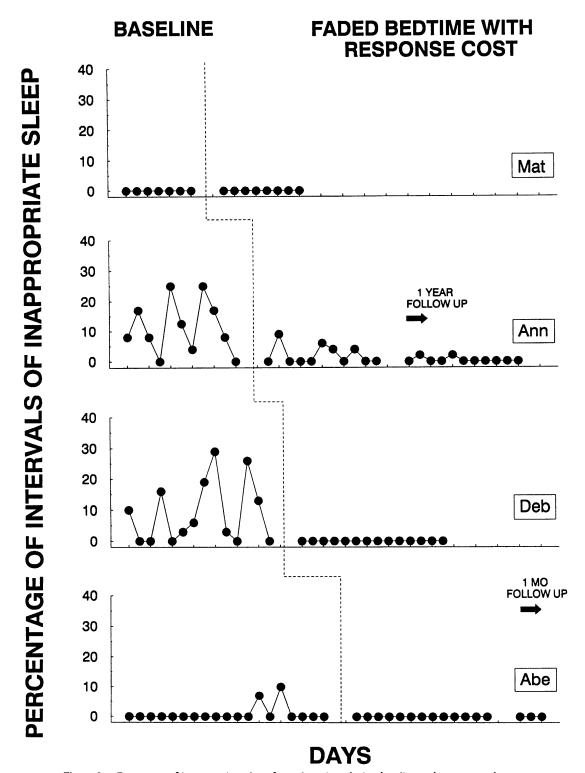


Figure 2. Percentage of inappropriate sleep for each patient during baseline and treatment phases.

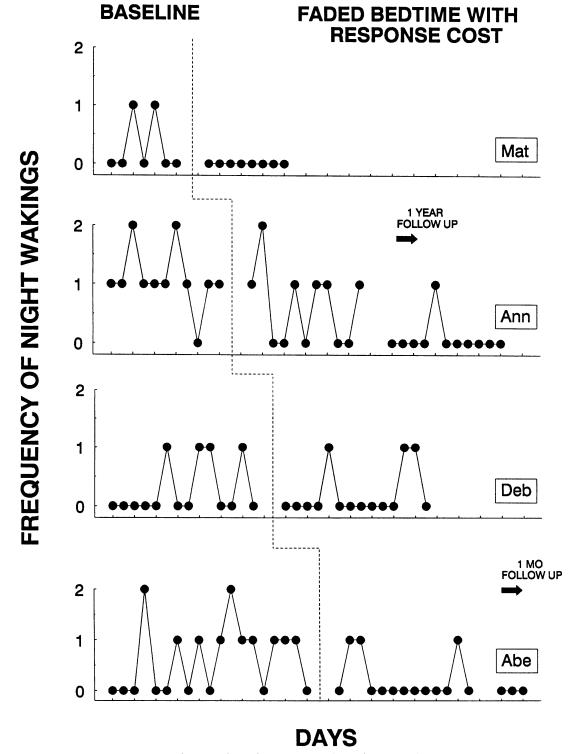


Figure 3. Frequency of night wakings for each patient during baseline and treatment phases.

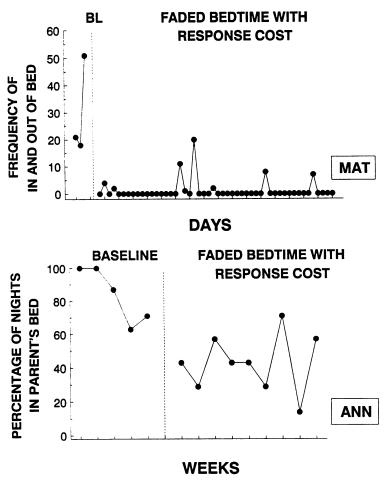


Figure 4. Frequency of in-and-out-of-bed behavior for Mat (top graph). Collateral effects of treatment: percentage of nights during the week that Ann was brought to her parents' bed (bottom graph).

DISCUSSION

This study is unique in that a single-subject experimental analysis, including 24-hr direct observation, was used to evaluate the effectiveness of the faded bedtime with response cost protocol on longstanding, multiple sleep problems in 4 developmentally delayed individuals. Each patient benefited from this procedure. Overall durations of sleep increased substantially for Mat and Ann during treatment, and Deb and Abe experienced increases in the regularity of their sleep. This treatment was also effective in reducing the night wakings of Abe and Ann, and the inappropriate sleep of Ann and Deb was substantially reduced.

From a theoretical perspective, both classical and operant conditioning may have facilitated treatment efficacy. In a classical conditioning framework, the unconditioned stimulus is the physiological state associated with sleep deprivation produced by setting the initial bedtime later than the average time of sleep onset during baseline. Ostensibly, the unconditioned response would be sleep. Because the children were only allowed to lie in bed for 15-min trials at times when they are likely to be tired, the protocol may increase the likelihood that being in a prone position in bed will become a conditioned stimulus that increases the probability of the sleep response.

From an operant standpoint, the procedure may

have an avoidance component (i.e., removal from bed) for long latency to sleep onset, which is hypothesized to function as follows. During baseline, the child had the opportunity to leave the bed, to have access to preferred activities and items or parental attention, and to return to bed when tired. During treatment, the child was required to leave the bed if not asleep within 15 min. While out of bed, the child had access to the same items and attention but could not return to bed until the next scheduled bedtime. As the child became more tired. the potency of the response cost (being removed from bed) may have increased, despite the availability of preferred items and/or attention. The child could avoid this potentially unpleasant event by displaying behaviors compatible with rapid sleep onset (e.g., lying quietly in bed with closed eyes). Finally, the procedure of fading allowed a stimulus, in this case bedtime, to be changed gradually such that the individual continued to respond to the changed stimulus (earlier bedtime) as he or she did to the original stimulus (late bedtime).

In addition to the behavioral procedures responsible for treatment efficacy, the protocol also capitalizes on several biological aspects of sleep. One biological factor that may facilitate treatment effects is the increase in sleep pressure that occurs when children are put to bed later than their average time of sleep onset during baseline. Increasing sleep pressure should increase the probability of rapid sleep onset and decrease the probability of night wakings (Dinges, 1986). A second biological factor that may operate with this intervention is that establishing a regular sleep-wake time may allow the body's circadian rhythms to become synchronized with the prescribed sleep-wake schedule. Regulation of sleep-wake times in accordance with a more normal circadian cycle may increase the probability that sleep patterns will be maintained when bedtime has been faded to an earlier hour and the manipulation of sleep pressure is no longer a factor. Finally, Mullaney, Johnson, Naitoh, Friedmann, and Globus (1977) have demonstrated that durations of sleep may be maintained at decreased amounts following gradual sleep reductions. Our data suggest that increases in sleep duration can

also be maintained over time through fading or by gradually increasing the length of sleep (e.g., for at least up to 1 year).

It is possible that the efficacy of the procedure lies in eliminating daytime sleep and that the treatment manipulations occurring at night are unnecessary. However, this does not account for the improvement in the sleep patterns of the 2 individuals who did not have excessive daytime sleep. Furthermore, Mullaney et al. (1977) have demonstrated that decreases in sleep during one period of the 24-hr day do not necessarily result in increases in sleep at another time of day. Thus, simply eliminating daytime sleep may not necessarily result in improved nighttime sleep.

Although the goal of many behavioral treatments is to effect large increases or decreases in behavior, the goal of treatment in sleep disorders is often the regulation of sleep patterns, requiring that the individual initiate the behavior (sleep) at about the same time every day. The behavior then persists for a relatively long period of time without stopping, ends at approximately the same time every day, and does not occur again for another long period of time. The current protocol helped to produce this behavioral pattern and appeared to have beneficial collateral effects.

Anecdotally, parents of the individuals in this investigation reported a high degree of satisfaction with treatment outcome. Reduction of night wakings decreased the need for nighttime parental supervision, resulting in increases in the parents' sleep durations. Several caretakers reported that children who had refused to go to bed in the past began requesting to go to bed (i.e., by pointing or gesturing) after discharge from the hospital. For 1 child, the need to bring her into the parents' bed was decreased, thus reducing a source of disruption in the family and discord in the parents' marriage.

Future research on the faded bedtime with response cost protocol should focus on determining its generality and limitations. One limitation of the current study is the specificity of the population treated—profoundly mentally retarded individuals with severe behavior problems. Further research is needed to determine the extent to which the pro-

cedure works with differing clinical populations with sleep disturbance as a feature (e.g., AD-HD, Tourette disorder), as well as with children who have insomnia without cooccurring behavior disorders.

A second potential limitation is that 3 of the 4 patients were treated on an inpatient unit with highly trained staff under careful supervision. The parents of the 4th child, who was treated as an outpatient, were well educated and highly motivated to change their child's sleep patterns. It is important to determine whether parents and caregivers differ in the extent to which they can accurately implement this protocol. Another possible limitation is the nonconcurrent multiple baseline design employed in the current investigation. However, the consistency of results across subjects renders the findings credible, which is the purpose of the design (Baer, Wolf, & Risley, 1987).

Because this treatment is multicomponent (i.e., fading and response cost), it would be of interest to conduct a component analysis to determine the relative contribution of the various elements of the procedure. Our initial study (Piazza & Fisher, 1989) involved fading the bedtime in 15-min increments and did not use a response cost component. Since that time, we have added the response cost component and have begun fading in 30-min increments (primarily for purposes of expediency). Based on clinical impressions, we believe the response cost is a useful component of treatment, but we have yet to test the validity of this hypothesis. We also believe that it may be better to begin fading in 30min increments, but to switch to 15-min increments if a patient reaches a plateau in the fading process where he or she does not show progress toward the final target bedtime and/or when the patient begins to approach his or her target bedtime. A component analysis would help to define the essential components of this treatment package.

Finally, to study whether regulation of circadian rhythms is related to the efficacy of the protocol, it would be useful to monitor changes in circadian rhythms by measuring body temperature on the same 24-hr schedule as we measured sleep. We could then determine whether circadian rhythms tend to be irregular during baseline and stabilize

following the initiation of treatment. It would be of particular interest to determine whether patients for whom the fading process is difficult show related difficulties in the regulation of circadian rhythms as measured by body temperature. It may be possible to improve the effectiveness of the intervention by using information about circadian patterns to set and adjust (i.e., fade) the child's bedtime.

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